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CASE REPORT | LIVER

Ischemic Cholangiopathy 11 Years after Liver Transplantation from Asymptomatic Chronic Hepatic Artery Thrombosis

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ABSTRACT

Hepatic artery thrombosis is a concerning complication of orthotopic liver transplantation, and it most often occurs early in the posttransplant period. However, on rare occasions it can occur at a time remote from transplant. We present a case of ischemic cholangiopathy complicated by stricture and anastomotic bile leak from chronic hepatic artery thrombosis that occurred 11 years after the transplant. The initial biliary stenting helped with the resolution of the leak but she was found to have stones, sludge and copious pus at the time of stent exchange. Hepatic arteriography demonstrated complete occlusion of the transplant hepatic artery with periportal collaterals reconstituting intrahepatic hepatic arterial branches. The patient was subsequently referred for repeat liver transplantation.

INTRODUCTION

Orthotopic liver transplantation has become the standard of care for acute and chronic liver failure. While advances in both surgical and medical modalities continue to improve outcomes, both short- and long-term complications can occur. One of the most concerning complications is hepatic artery thrombosis (HAT), which is the most common complication necessitating repeat transplanation.[1] HAT is of greatest concern in the immediate transplant setting because it often presents with ischemia and necrosis. Early and late HAT are differentiated by a cutoff of 21 days.[2] Late HAT is less clinically conspicuous although still serious, most often presenting with biliary pathology with a median time to presenation of 4–6 months after transplant.[3-5] Over time, the incidence of HAT decreases, rarely occuring more than 5 years after transplant. However, patients remain at risk for the lifetime of the graft.

CASE REPORT

A 54-year-old woman with a history of well-controlled diabetes mellitus, tobacco abuse, and autoimmune hepatitis status post orthotopic liver transplant (OLT) 11 years prior was found on routine follow-up to have asymptomatic elevation in her liver tests: alkaline phosphatase 313 U/L (normal 25–125 U/L), alanine aminotransferase 92 U/L (normal 7–52 U/L), and total bilirubin 2.7 mg/dL (normal 0–1.0 mg/dL). Her posttransplant course had previosuly been benign outside of recurrent autoimmune hepatitis 8 years prior, which responded to steroids. No changes were made to her immunosuppressive regimen. She denied recent illnesses, alcohol, or illicit drugs, and she reported compliance with her medications. Evaluation with magnetic resonance cholangiopancreatography revealed severe donor-duct biliary dilation, anastomotic stricture, and periductal cystic changes with normal recipient common duct and pancreatic duct caliber (Figure 1). Endoscopic retrograde cholangiopancreatography (ERCP) revealed a moderate biliary stricture at the posttransplant anastamosis, with marked dilation of the donor biliary tree and anastomotic bile leak concerning for ischemic cholangiopathy. No stones were noted, but they were not

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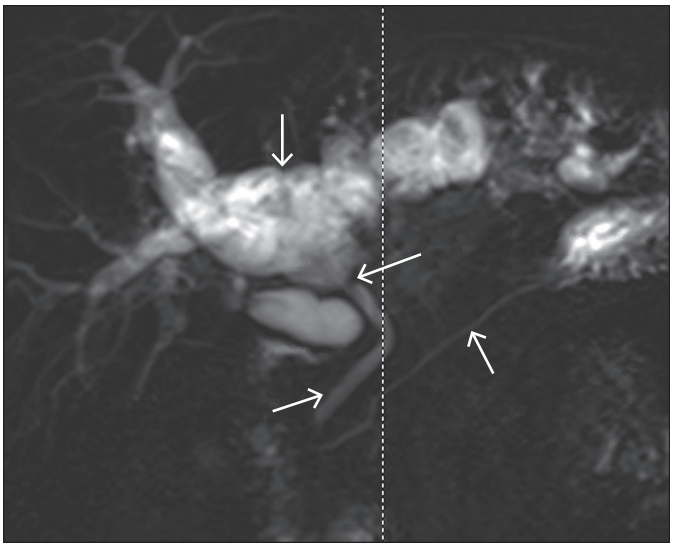

Figure 1. Magnetic resonance cholangiopancreatography image showing a normal-caliber common bile duct, a pancreatic duct with stricture, and massive intrahepatic biliary duct dilation.

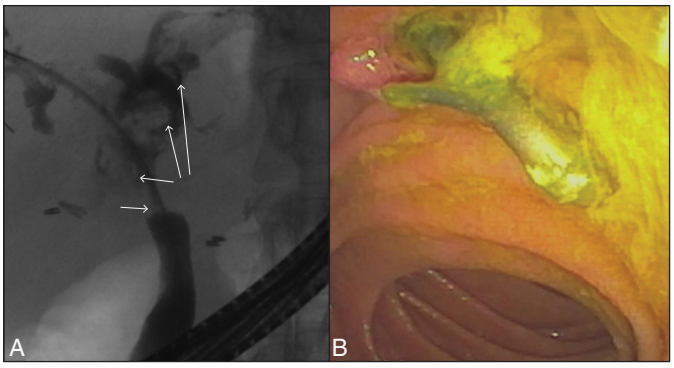

Figure 2. (A) Fluoroscopic image of cholangiogram with biliary stricture and stones. (B) Endoscopic image of pus and sludge draining from common bile duct stent.

specifically addressed in view of the leak. An 8.5-Fr biliary stent was placed into the donor duct with a repeat ERCP planned at 2 months follow-up. Computed tomography with contrast was negative for biloma or mass, but visualization of the hepatic artery was poor. The patient was monitored overnight and discharged with significant clinical improvement.

Six weeks later she presented with complaints of malaise, nausea, and anorexia, but her evaluation showed normal liver tests and minimally impaired synthetic function (international normalized ratio 1.38). Repeat ERCP for stent exchange showed severe dilation of the donor ducts with stones, sludge, and copius pus (Figure 2). The leak had resolved. This raised concern for the integrity of the hepatic artery and ischemic cholangiopathy. Hepatic arteriography demonstrated complete occlusion of the transplant hepatic artery with periportal collaterals reconstituting intrahepatic hepatic arterial branches (Figure 3). The presence of collateral circulation and the patient's asymptomatic clinical presentation suggested that she had developed chronic hepatic artery thrombosis (HAT) at a time remote from transplantation, which led to chronic ischemic cholangiopathy and bile duct disruption with a leak. Interventional radiology did not feel the lesion was amenable to intravascular intervention. After discussion with Transplant Surgery, evaluation for repeat transplantation was deemed the most appropriate course of action.

DISCUSSION

This case represents rare timing of a dangerous complication of OLT with classic clinical, radiographic, and endoscopic findings. HAT can be seen in up to 9% of OLT patients,[2] and while acute HAT is concerning due to its dramatic presentation, late HAT has its own serious consequences. Given the necessary severing of natural capsular collaterals with OLT, the biliary tree is dependent on the hepatic artery, leading to

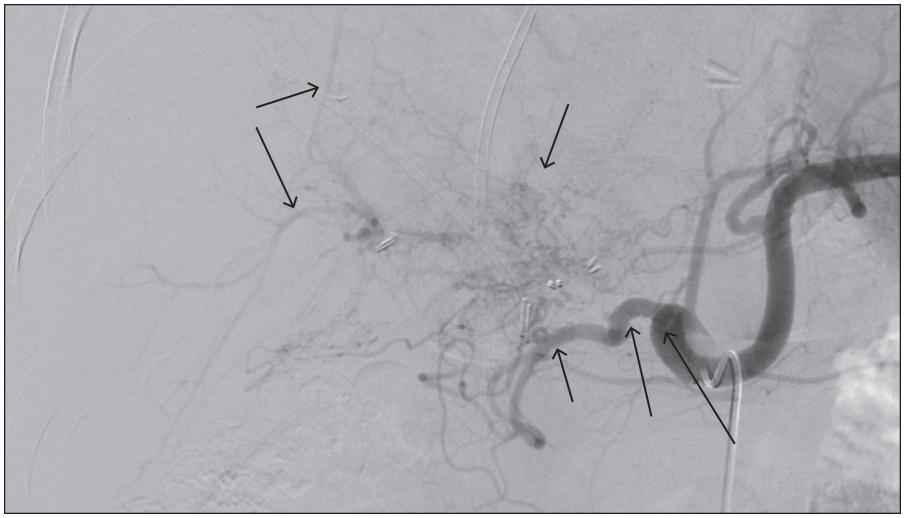

Figure 3. Angiographic image of the celiac artery showing the abrupt termination of common hepatic artery with significant collateralization arising from the gastroduodenal artery.

the risk of ischemic cholangiopathy with hepatic artery compromise. Smoking and diabetes, as seen in this case, may increase the risk for HAT, but the underlying cause is often unclear. This ischemia leads to biliary strictures, leaks, sludge, stones, and bile duct casts, all of which can be complicated by recurrent cholangitis.[6] Nonsurgical approaches to management, including percutaneous stricture dilation, stenting, and repeat therapeutic ERCP, have become the standard of care in these patients. However, patients who have failed these therapies or who present with recurrent cholangitis require repeat transplantation.[7,8] This case is instructive with regard to both basic hepatic vascular and biliary anatomy and the need for adequate understanding of clinical hepatology, transplant hepatology, and advanced endoscopy. Furthermore, it is imperative for the general gastroenterologist to recognize this entity because medical advances have led to longer complication-free posttransplant survival times. As a result, more of these patients will return to the general gastroenterology practice for longitudinal care.

DISCLOSURES

Author contributions: E. Krajicek and R. Vuppalanchi wrote the manuscript. S. Sherman, M. Lacerda, and M.S. Johnson edited manuscript. R. Vuppalanchi is the article guarantor.

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